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Parent-mediated communication interventions for improving the communication skills of preschool children with non-progressive motor disorders (Protocol)

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Parent-mediated communication interventions for improving the communication skills of preschool children with non-progressive motor disorders

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ABSTRACT

This is a protocol for a Cochrane Review (Intervention). The objectives are as follows:

To assess the effectiveness of parent-mediated communication interventions in improving the communication skills of preschool children with non-progressive motor disorders.

Specifically, this review aims to determine the effectiveness of parent-mediated communication interventions in improving the communication of preschool children with non-progressive motor disorders, when compared to no intervention, and when compared to clinician-mediated interventions.

BACKGROUND

Description of the condition

Non-progressive motor disorders in childhood arise from a variety of conditions, including cerebral palsy, acquired brain injury, global developmental delay, Down syndrome and genetic mutations. Exactly how many children are affected is currently unknown due to sparse population-level data. The most comprehensive data come from international surveillance of cerebral palsy. Cerebral palsy is defined as “a group of permanent disorders of

the development of movement or posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain” (Rosenbaum 2007). Registries have shown that cerebral palsy affects 2 to 3 per 1000 children in high-income countries (Cans 2008; Kirby 2011; Reid 2011). Prevalence is likely to be greater in low- and middle-income countries where health care is less abundant, but cerebral palsy rates in these regions have not yet been ascertained. A recent Dutch study estimated that in children under 14 years of age there are about 3.6 new cases of severe acquired brain injury per 100,000 children per annum (de Kloet 2013). However, it

is unclear how many of these children have ongoing motor disorders. Again, the incidence may be greater in low- and middle-income countries with less access to public health services. Global developmental delay and Down syndrome lead to low motor tone and slow acquisition of motor skills, and affect approximately 39 and 1.4 per 1000 children respectively (Boyle 2011; Parker 2010). Genetic mutations that cause motor disorders include the PRRT2 mutation and the GLUT-1 syndrome (Blackburn 2012), but their prevalence is unclear. Many developmental disabilities, including those causing non-progressive motor disorders included in this review, are more common in boys and in families in who live in poverty (Boyle 2011). Disorders are diagnosed by paediatricians, paediatric neurologists and geneticists. Differential diagnosis may take some years due to the slowly evolving nature of some conditions.

Motor disorders impair the range, speed, strength and consistency of movements. When disorders affect the movements underpinning vocalisation, speech, gesture and/or facial expression, parents and other caregivers find it hard to recognise and interpret children's attempts to communicate and this can lead to interaction breaking down (Hanzlik 1990; Light 1985; Pennington 2001). To promote effective interaction, parents may structure conversations around the children's communication signals that are easy to understand (Dunst 1985; Tannock 1992). However, this can lead to asymmetrical interaction, with parents introducing topics, asking forced choice questions and then acknowledging their child's response. Such an uneven, parent-led pattern of conversation can make it difficult for children with motor disorders to learn new communication skills.

It is estimated that around 22% of children with cerebral palsy have speech intelligibility limitations due to the motor impairments and a further 20% to 30% have no functional speech (Nordberg 2013; Parkes 2010; Stanley 2000). Speech disorder is more likely to occur in dyskinetic and ataxic forms of cerebral palsy than in spastic cerebral palsy (Bax 2006; Parkes 2010), and is more common in bilateral than unilateral distribution in spastic type (Parkes 2010). The prevalence of speech disorders in other conditions leading to non-progressive motor disorders is currently unknown.

Children with motor disorders who also have a cognitive impairment may take longer to reach milestones, such as intentionality and engaging in joint attention with another person, which are vital for interaction, and the development of linguistic understanding may be delayed. Approximately half (49%) of children with cerebral palsy have an intellectual disability (IQ less than 70) and 28% have a severe intellectual disability (IQ less than 50) (Novak 2012). Current research suggests that receptive language is largely commensurate with cognitive development in cerebral palsy (Pirila 2007) but further epidemiological studies are needed to confirm this.

Communication difficulties have a profound impact on children's family, social and educational life. Children with communication and motor disorders are more at risk of lower quality of life and

restricted social participation than their peers with and without motor disorders (Dickinson 2007; Fauconnier 2009). The impact of communication breakdown is felt throughout families, and parents report high levels of stress (Parkes 2011; Pousada 2013).

As differential diagnosis may not be possible in early childhood and all motor disorders affecting speech and gesture can lead to intelligibility limitations, this review will be inclusive of all causes of non-progressive motor disorders in the preschool years. One exception to this is Down syndrome. A separate review will consider communication interventions for parents of children with Down syndrome (O'Toole 2016). Therefore, we will exclude studies examining only children with Down syndrome, but will include studies in which Down syndrome is one of a range of disorders causing motor impairment. Degenerative disorders, such as muscular dystrophies, and metabolic disorders may also be associated with motor impairment, and may become apparent after a period of healthy development. As these disorders lead to a loss of skills rather than development following an atypical pattern, as is the case for children with non-progressive disorders, they will not be included in this review. Also, children with severe hearing or visual impairments, or both, have specific difficulties acquiring early interaction skills arising from their differences in processing communication signals, which are beyond the scope of this review.

Description of the intervention

As communication skills are developed in interaction, and children's most frequent communication partners are their parents, therapy involves training parents to adapt their communication style. This is referred to as parent-mediated or indirect therapy. It aims to help parents of children with motor disorders to recognise and interpret their child's attempts to communicate and to stimulate their child's development of new skills (e.g. Bruno 1998; Girolametto 1986; Kaiser 1987; Kent-Walsh 2015; Mahoney 1988; Pepper 2004; Ronski 2010; Yoder 2002). Training is most often provided by speech and language therapists and other personnel with an interest in interaction (e.g. psychologists and early-years educators). Training can be delivered to individuals or groups of parents and may take place in parents' homes or in health, education or social care settings.

Training often teaches parents about how communication develops, from pre-intentional, reflexive communication through to nonverbal, intentional communication and then on to linguistic communication (Hemmeter 1994; Pepper 2004). It covers the purposes for which communication is used and how communication involves communication partners taking turns in expressing and receiving signals (Hemmeter 1994; Mahoney 1988; Pepper 2004). Techniques to aid communication development are introduced, including creating stimulating environments, promoting a need to communicate, allowing sufficient time for children to enter or start conversation, and responding contingently to children's messages. Parents are encouraged to apply this information

to their own interaction with their child (Fey 2006; Gibbard 2004; Kaiser 2001; Mahoney 1988; Pepper 2004). Training often includes coaching, whereby therapists watch the interaction between the parent and child, in real time or on video, and highlight which behaviours prompt the child to communicate so the parent can repeat these more frequently (Kaiser 1995; Kaiser 2003; McDuffie 2016; Pepper 2004). It might also involve the parent watching the therapist modelling interaction with the child (Kaiser 2003; Pepper 2004).

Young children who have severe speech impairment associated with their motor disorders may be introduced to augmentative and alternative communication (AAC) to supplement their natural forms of communication. AAC includes signing and use of body movements (unaided AAC), or may introduce equipment such as objects to represent daily activities, photographs, pictures, symbols and speech generating devices (aided AAC). AAC provides access to a wider range of vocabulary and language, but does take time to learn by children and their parents. Training is often provided for parents to teach them how to accommodate the use of the system in conversation and help their children to produce new vocabulary and language structures via the AAC system (Kent-Walsh 2015). Such training may be incorporated in the generic communication training described above (Pennington 2009) or be provided separately in programmes that focus specifically on AAC (Kent-Walsh 2015).

How the intervention might work

Parent communication training is based on the transactional theory of development, which hypothesises that children and their parents continuously adapt to each other's behaviours (Sameroff 2000). Following this hypothesis, helping parents to recognise and interpret their child's current communication behaviours and adapt their own interaction style to accommodate their child's physical limitations and create more frequent and appealing opportunities for the child to communicate, should enable parents to prompt their child to communicate more frequently using any intelligible mode (e.g. vocalisation, speech, gesture, AAC). Teaching parents about how communication develops should also enable them to continue to stimulate their child's development by prompting the use of communication for a wider range of purposes and scaffolding the production of more sophisticated communication signals and the use of a wider vocabulary (Girolametto 1996). Changes in parents' conversation behaviours include: giving their children more time to start interactions and produce messages, responding contingently to children's communication, taking shorter turns in conversation and using less complicated language. Such changes should prompt children to take more turns in interaction, initiate conversation more frequently, and use communication for a greater range of purposes with a wider range of vocabulary. The intervention may also serve to increase parents' confidence in their communication with their children, re-

duce parental stress as communication breakdowns become less frequent, and help children to interact successfully in a greater number of social activities and with a broader range of people.

Why it is important to do this review

The timing and intensities of interventions, and the effectiveness of communication interventions were rated as the two most important areas for investigation in a recent James Lind Alliance Childhood Disability Research Priority Setting Partnership (Morris 2015). Internationally, there has been a drive in research to develop early interventions to maximise the potential skill development associated with brain plasticity in infancy and the early years. Early communication intervention has often focused on training parents as children's most frequent communication partners, and parent training is now routinely provided by speech and language therapists to families of preschool children with motor disorders (Watson 2015). Previous Cochrane reviews have considered parent training programmes for children with autism (Oono 2013) and primary speech and language delay or disorder (Law 2003), and a future review will investigate parent training for children with Down syndrome (O'Toole 2016). However, the method of delivery of parent training, its contents, dosage and suitability for families of children with motor disorders, have not been evaluated recently. A previous review considered speech and language therapy interventions to improve the communication skills of children with cerebral palsy and included parent-mediated interventions (Pennington 2003); its authors identified one randomised controlled trial of a parent training communication intervention. This review will update the section of the previous review that examined training delivered to parents of children with cerebral palsy (Pennington 2003), to identify new empirical data. It will also consider intervention provided to parents of preschool children with other non-progressive motor disorders, as their communication development is similarly affected. Including all children with non-progressive motor disorders will enable examination of the generic effectiveness of parent training interventions in the preschool period, extending the utility of the review to service providers and policy makers.

OBJECTIVES

To assess the effectiveness of parent-mediated communication interventions in improving the communication skills of preschool children with non-progressive motor disorders.

Specifically, this review aims to determine the effectiveness of parent-mediated communication interventions in improving the communication of preschool children with non-progressive motor disorders, when compared to no intervention, and when compared to clinician-mediated interventions.

METHODS

Criteria for considering studies for this review

Types of studies

Randomised controlled trials (RCTs) including cluster-RCTs, and quasi-RCTs in which participants are allocated to intervention groups by methods that are not strictly random.

Types of participants

Children up to five years of age who have a communication difficulty associated with any non-progressive motor disorder acquired before two years of age. We will include children with additional intellectual impairments, including children with Down syndrome, if they have identified motor difficulties. We will exclude studies of only children with Down syndrome, as they will be considered by the review [O'Toole 2016](#), and studies of children whose vision is corrected by spectacles and whose hearing is amplified by hearing aid(s). We will exclude children whose communication is primarily limited by a sensory impairment, as their communication development differs from children who can see and hear the world around them. We will infer motor disorder from descriptions of children's development and confirm this with study authors, if necessary. Communication difficulty will be diagnosed by speech and language therapists or psychologists. Parents of the children above.

Types of interventions

We will consider studies of training delivered to parents with the aim of helping them to promote their child's communication development. Training can be delivered to parents individually or in groups. Training can be delivered by speech and language therapists, psychologists or early educators. Training can take place in the home or in health, education or community support settings. Training programmes may vary in dosage: intensity, frequency and duration. Training may include communication via augmentative and alternative communication (AAC) as one mode of communication in a total communication approach or may focus primarily on communication using AAC. We will exclude facilitated communication.

Comparisons of interest are training delivered to parents versus treatment as usual (e.g. multidisciplinary therapy groups providing motor, sensory and language stimulation); parent training versus clinician-mediated intervention and parent training versus no intervention or waiting-list controls.

Types of outcome measures

Primary outcomes

1. Children's ability to communicate effectively in everyday life. Outcomes include children's ability to:
 - i) take turns in conversation, initiating conversation and responding to others' conversational gambits;
 - ii) use communication for a wide range of purposes such as requesting attention, asking questions, answering questions, making comments and repairing conversation when they have not been understood; and
 - iii) use a range of modes of expression by vocalising, speaking, using gesture or using the AAC system.
2. Adverse events, including reductions in the frequency with which children communicate, or increases in negative behaviour. Outcomes will be measured at the level of activity (i.e. the ability to execute a task), and at the level of participation (i.e. communication in life situations) ([WHO 2001](#)). Measures will include rating scales (e.g. Focus on the Outcomes of Communication Under Six ([Thomas-Stonell 2010](#)) and Therapy Outcome Measures ([Enderby 2015](#))), communication assessments (e.g. Communication and Symbolic Behavior Scales ([Wetherby 2002](#))), and observational coding schemes (e.g. frequency counts of children's initiations and responses in interaction).

Secondary outcomes

1. Child outcomes:
 - i) speech and language function, as assessed using standardised measures of children's expressive and receptive language skills and speech production (e.g. Pre-school Language Scales ([Zimmerman 2002](#)); Communicative Development Inventory (CDI; [Fenson 2006](#)); Receptive-Expressive Emergent Language Scale-3 (REEL-3; [Bzoch 1970](#))); non-standardised assessments of gestural ability; or production of messages using AAC on demand, as measured using coding schemes developed for individual research studies that include validity and reliability data; and
 - ii) children's generic participation, as assessed using validated measures such as Assessment of Life Habits ([Noreau 2007](#)), and Children's Assessment of Participation and Enjoyment ([King 2004](#)).
2. Parent outcomes:
 - i) parents' communication and interaction strategies, as assessed using non-standardised measures such as Responsive Augmentative and Alternative Communication Style ([Broberg 2012](#)); coding schemes, which measure the frequency of parent communication behaviours (e.g. initiations of conversation; directives) developed for individual research studies that include validity and reliability data;

- ii) family stress and coping (e.g. Questionnaire on Resources and Stress (Friedrich 1983) or Carer Strain Index (Robinson 1983));
- iii) satisfaction of patient and family with treatment (e.g. rating scales developed for individual studies, Patient Satisfaction Questionnaire Short Form (PSQ-18; Marshall 1994)); and
- iv) compliance with treatment (e.g. number of sessions missed and reasons for this).

We will compare baseline measures with outcomes grouped into the following time points: short term (zero to one month following intervention completion), medium term (two to five months after intervention) and long term (six or more months following intervention).

We will combine results from studies where tools measure the same outcome using the same type of data (e.g. frequency of child communication behaviours; standard scores on child language measures).

Search methods for identification of studies

Electronic searches

We will search the electronic databases and trials registers listed below, from inception onwards.

1. Cochrane Central Register of Controlled Trials (CENTRAL; current issue) in the Cochrane Library, and which includes the Cochrane Developmental, Psychosocial and Learning Problems Specialised Register.
2. MEDLINE Ovid (1946 onwards).
3. CINAHL Plus EBSCOhost (Cumulative Index to Nursing and Allied Health Literature; 1937 onwards).
4. Embase Ovid (1974 onwards).
5. PsycINFO Ovid (1806 onwards).
6. Science Citation Index Web of Science (SCI; 1970 onwards).
7. Conference Proceedings Citation Index - Science Web of Science (CPCI-S; 1990 onwards).
8. Language and Linguistic Behaviour Abstracts ProQuest (LLBA; 1871 onwards).
9. British Education Index EBSCOhost (BEI; 1929 onwards).
10. ERIC EBSCOhost (Education Resources Information Center; 1966 onwards).
11. *Cochrane Database of Systematic Reviews* (CDSR; current issue) in the Cochrane Library.
12. LILACS (lilacs.bvsalud.org/en).
13. National Rehabilitation Information Center (naric.com).
14. SpeechBITE (speechbite.com).
15. ClinicalTrials.gov (clinicaltrials.gov).
16. EU Clinical Trials Register (www.clinicaltrialsregister.eu/ctr-search/search).

17. NIH Clinical Research Center (www.cc.nih.gov/home/clinicalstudies.html).

18. UK Clinical Trials Gateway (www.ukctg.nihr.ac.uk/clinical-trials).

19. World Health Organisation International Clinical Trials Registry Platform (WHO ICTRP; www.who.int/ictip/en). We will use the search strategy in [Appendix 1](#) to search Ovid MEDLINE. We will adapt the search appropriately for other databases. We will not limit the search by the country in which the research was undertaken, the language in which the research is reported, year of publication or publication status. We will seek translations of papers published in languages other than English when necessary.

Searching other resources

We will handsearch the reference lists of relevant papers and reviews for studies not identified by the electronic searches. We will approach authors working in the field to locate currently unpublished studies.

Data collection and analysis

Selection of studies

One review author (KL) will conduct all searches. Two authors (LP and KL) will independently screen each title and abstract for eligibility against the inclusion criteria (see [Criteria for considering studies for this review](#)). When inclusion is uncertain we will obtain the full text of the paper. Two of the three review authors (LP, JG or KL) will be randomly allocated to each paper that appears from the abstract to fit the inclusion criteria and will independently review each paper to determine its inclusion. In the event of disagreement regarding inclusion, the third review author (LP, KL or JG) will review the paper independently and we will reach consensus through discussion and by reassessing the inclusion criteria together. We will record our decisions in a study flow diagram (Moher 2009).

Data extraction and management

We will develop a tool for extracting data for this review. All authors will be involved in data extraction. Two members of the research team (LP, JG or KL) will be randomly assigned to each paper and will independently extract data into Review Manager (RevMan) version 5 (RevMan 2014). We will resolve disagreements by discussion and by involving the third author (LP, JG or KL). We will collect the following data.

1. Country of origin.
2. Type of study: RCT, cluster-randomised trial, quasi-RCT.
3. Sample size: treatment and control groups, attrition.

4. Study population: parents (age, gender, relationship to child, educational level (high school, further education, higher education)); children (diagnosis of underlying disorder, type of motor disorder (spastic, dyskinetic, ataxic, hypotonic, mixed); age; gender; non-verbal cognitive development (standard scores, percentile rank); receptive language development (standard scores, percentile rank); modes of communication used (vocalisation, speech, gesture, facial expression, body movement, AAC); communicative functions used; number of intelligible words; gross motor function, as classified using the Gross Motor Function Classification System (GMFCS; [Palisano 2007](#)); and upper limb function, as categorised using the Manual Ability Classification System (MACS; [Eliasson 2006](#)), when possible.

5. Intervention: type of intervention; duration; frequency of sessions; group or individual; content of sessions; inclusion of coaching or didactic teaching only.

6. Comparator intervention: type of intervention; duration; frequency of sessions; group or individual; content of sessions; inclusion of coaching or didactic teaching only.

7. Intervention provider: speech and language therapist (or relevant term in country of origin), psychologist, teacher, other.

8. Fidelity of intervention: how this was assessed and by whom.

9. Outcome measures: parent outcomes; child outcomes; family outcomes.

10. Results: short term (zero to one month following intervention completion), medium term (two to five months after intervention) and long term (six or more months following intervention)

11. Adverse effects.

12. Conflicts of interest, including declarations of conflicts of interest.

Assessment of risk of bias in included studies

We will extract information on each study about risk of bias. We will rate the risk of bias using the Cochrane 'Risk of bias' tool ([Higgins 2011a](#)). Two of the three review authors (LP, KL or JG) will be randomly allocated to each study to extract data and rate risk of bias. Any disagreements will be resolved through discussion or by involving the third review author (LP, KL or JG, i.e. author not assigned to the paper under review). We will rate studies as having low, high or unclear risk of bias in: random sequence generation; allocation concealment; blinding of participants and personnel; blinding of outcome assessment; completeness of data collection; selective reporting; and other sources of bias. We will apply the coding schedule in [Appendix 2](#) for each source of bias.

Measures of treatment effect

Binary data

It is possible that some studies may present binary data (e.g. treatment effect achieved or not achieved). For such studies we will calculate an odds ratio (OR) with a 95% confidence interval (CI).

Continuous data

We expect most studies to measure intervention success using continuous measures (e.g. standardised tests of speech and language, number of intelligible words, number of communicative functions, frequency of communication). When studies have used the same continuous outcome measure, we will report the effect size as a mean difference (MD), with 95% CI. For studies that evaluate the same construct using different continuous outcome measures that share the same method of administration (e.g. questionnaires; frequency counts of behaviours measured in direct observation), we will summarise results using the standardised mean difference (SMD) with 95% CI.

Unit of analysis issues

Cluster-randomised trials

Cluster-randomised trials may be retrieved in the review; for example, service providers may be allocated to provide a specific type of intervention. If we identify cluster-randomised trials we will follow the guidance in the *Cochrane Handbook for Systematic Reviews of Interventions* on managing such data ([Higgins 2011b](#), section 16.3). We will check that appropriate analyses have been undertaken (e.g. two sample t-tests comparing the means of the clusters in the intervention group at cluster level or mixed-effect linear regression using individual participant data ([Donner 2000](#))). If this is not certain, we will seek to extract or calculate effect estimates and their standard errors and adjust the standard errors to account for clustering ([Donner 1980](#)). Adjustment will require intraclass correlation coefficients (ICC) to be reported ([Donner 1980](#)). If ICCs are not in the published reports we will request them from study authors. If ICCs are not available we will search similar studies to obtain external estimates of the ICC for the relevant outcomes. If no external estimates are available we will undertake sensitivity analyses using a high ICC of 0.100, a moderate ICC of 0.010 and a small ICC of 0.001 (see [Sensitivity analysis](#)). Following [Higgins 2011b](#) (section 16.3.6), we will obtain standard errors that account for clustering by multiplying the standard errors of the effect estimate by the square root of the design effect. We will combine the estimates and adjusted standard errors from cluster-randomised trials with those from trials allocating individual participants to groups, using the inverse variance method in [RevMan 2014](#), providing the groups of participants in the trials are similar ([Higgins 2011b](#), section 16.3.7).

Cross-over trials

It is possible that trials might compare parent training interventions (e.g. if comparing a method of delivery or the effects of a specific topic). In such trials we will include data from the first period only, so as not to count the same participant twice.

Studies with multiple treatment groups

We expect most studies to compare one type of parent-mediated intervention with no treatment or an intervention delivered by the therapist directly to the child. However, if a study investigates multiple treatment groups, we will make single pair-wise comparisons by combining data from all eligible parent training intervention groups and comparing these with data combined from all eligible control groups, as recommended in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011b, section 16.5.4).

Dealing with missing data

We will request missing data from study authors and send two reminder emails one month apart. We will specifically request data on outcomes and reasons for withdrawals from the study. We will describe missing data and the resulting potential bias using the 'Risk of bias' tool and will note this risk of bias in the Results section of the review. We will refer to the *Cochrane Handbook for Systematic Reviews of Interventions* for methods of dealing with missing data (Higgins 2011b, section 16.1). If the authors undertook an intention-to-treat analysis, we will use all the results provided. If an intention-to-treat analysis was not undertaken, and continuous data are considered missing at random, we will impute data using a 'last case carried forward' analysis. If binary data are considered missing at random, we will undertake a sensitivity analysis, adopting both a best- and worst-case scenario in which, for example, children in the experimental group are imputed to have a good outcome and poor outcome respectively (see [Sensitivity analysis](#)). If binary data are considered not to be missing at random, we will impute the data assuming that the missing data would be negative (i.e. that data are missing because families dropped out of the study because of poor outcomes). If summary data that are required for meta-analysis (e.g. standard deviations) are not reported or provided by authors on request, we will derive them using the calculations provided in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011b, section 16.1.3). We will address the potential impact of missing data in the Discussion section of the review.

Assessment of heterogeneity

We will assess heterogeneity in clinical characteristics of study samples (e.g. parents' educational level or socioeconomic status, ratio of mothers to fathers in group composition; children's age, type

or distribution of motor disorder, level of intellectual impairment, receptive or expressive language, use of AAC) and trial characteristics (e.g. intervention duration and frequency, delivery to individuals or groups of parents, randomisation, concealment, blinding of outcome assessment, losses to follow-up). We will discuss any differences between studies in full. We will use the Chi^2 test to assess if statistical heterogeneity is likely to be due to chance alone. We will use the I^2 test and Tau^2 to describe the variation in effect estimates that is due to heterogeneity rather than sampling error (Higgins 2002).

Assessment of reporting biases

We will seek to minimise reporting bias in this review by searching all publication types, not limiting searches to English language and by contacting authors in the field. Should we identify more than 10 studies that fit the inclusion criteria we will use funnel plots of effect estimates to assess the possibility of publication bias on primary outcomes. We will use Egger's test to test for funnel plot asymmetry (Egger 1997).

Data synthesis

If there are two or more studies reporting interventions that are similar in terms of topic, delivery methods and dosage (duration, frequency and intensity of sessions), and that include similar participants (parents and children) and use similar outcome measures, we will undertake meta-analysis using [RevMan 2014](#), applying a random-effects model.

We expect most studies to use continuous measures. However, if an outcome is measured using binary data in some studies and continuous measures in others, we will convert binary results from an OR to a SMD if the continuous measure has an approximately normal distribution or logistic distribution. If data are not normally or logistically distributed, we will conduct separate analyses. We will calculate overall effects using inverse variance methods.

'Summary of findings' tables

We will assess the overall quality of the body of evidence for each outcome using the GRADE approach (GRADE 2008), and assign ratings of 'high', 'moderate', 'low' or 'very low' quality. As per the GRADE recommendations, the following five factors may reduce the quality level assigned: limitations in the design and implementation of available studies, which suggest a high likelihood of bias; indirectness of evidence (indirectness of population, intervention, control or outcomes); unexplained heterogeneity or inconsistency of results (including problems relating to subgroups); imprecision of results, as shown by wide CIs; and high probability of publication bias.

All review authors will be involved in the grading of evidence quality. Two review authors (LP, KL or JG) will be randomly assigned to an outcome and will independently assess the quality of the

body of evidence for that outcome. We will resolve disagreements by involvement of the third review author (i.e. author who is not assigned to the outcome). When a review author is an author of an included study they will not be involved in the assessment of evidence quality. We will use GRADEprofiler ([GRADEPro GDT 2015](#)) to import data from [RevMan 2014](#), to construct 'Summary of findings' tables. We will present all results for the primary outcomes (children's communication activity and communicative participation; adverse events) and secondary outcomes (children's speech and language function; children's generic participation; parents' communication and interaction; family stress and coping; satisfaction of patient and family with treatment; compliance with treatment) in separate 'Summary of findings' tables.

Subgroup analysis and investigation of heterogeneity

We will conduct subgroup analyses to explore possible sources of heterogeneity:

1. in the presence of severe or profound intellectual or receptive language impairment (impairment in either function more than or equal to -1.9 standard deviations versus nonverbal or receptive language score less than -2 standard deviations);
2. in parental education (high school versus further or higher education);
3. in dosage of intervention (frequency and duration of sessions); and
4. between specific 'named' interventions (e.g. Hanen programmes (see, for example, [Pepper 2004](#)) or Enhanced

Milieu Teaching (see, for example, [Hemmeter 1994](#))).

Sensitivity analysis

We will use our 'Risk of bias' assessment to inform sensitivity analyses. As it is difficult to blind parents and training providers to the type of intervention, sensitivity analyses will use data from risk of bias arising from random allocation generation, allocation concealment, loss to follow-up, and incomplete reporting of outcomes. We will remove studies judged to have a high risk of bias in these areas to determine their effect on the pooled estimate. We will undertake a sensitivity analysis of binary outcomes if data are considered missing at random, adopting both a best- and worst-case scenario in which, for example, children in the experimental group are imputed to have a good outcome and poor outcome respectively.

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* Indicates the major publication for the study

APPENDICES

Appendix I. Ovid MEDLINE search strategy

- 1 Cerebral Palsy/
- 2 cerebral pals\$.tw,kf.
- 3 (ataxic or ataxia\$).tw,kf.
- 4 spastic\$.tw,kf.
- 5 dyskinetic\$.tw,kf.
- 6 (non-progressive adj3 (disabilit\$ or disorder\$ or impair\$)).tw,kf.
- 7 Movement Disorders/
- 8 motor disorders/
- 9 motor skills disorders/
- 10 Motor Skills/
- 11 (movement adj3 (disorder\$ or disabilit\$ or impair\$)).tw,kf.
- 12 (motor adj3 (disorder\$ or disabilit\$ or impair\$)).tw,kf.
- 13 Brain Injuries/
- 14 Brain Damage, Chronic/
- 15 brain injury, chronic/
- 16 (acquired brain injur\$ or ABI).tw,kf.
- 17 Developmental disabilities/
- 18 (developmental\$ adj3 (delay\$ or disab\$)).ti,kf.
- 19 Down Syndrome/
- 20 down\$ syndrome\$.tw,kf.
- 21 dyskinesias/
- 22 Chorea/
- 23 (dyskenesia\$ or chorea\$).tw,kf.
- 24 Angelman Syndrome/
- 25 Angelman\$ syndrome\$.tw,kf.
- 26 (GLUT1 or GLUT-1).tw,kf.
- 27 (PRRT2 or PRRT-2).tw,kf.
- 28 or/1-27

29 Infant/
 30 exp child/
 31 (child\$ or infant\$ or babies or baby or toddler\$ or girl\$ or boy\$ or pre-school\$ or preschool\$ or nurser\$ or kindergarten\$ or kindergarten\$).tw,kf.
 32 or/29-31
 33 28 and 32
 34 exp Parent-Child Relations/
 35 exp Parents/
 36 Parenting/
 37 Family/
 38 (at home or (in adj3 home) or home-based).tw,kf.
 39 Caregivers/
 40 or/34-39
 41 education/
 42 teaching/
 43 "Early intervention (Education)"/
 44 early intervent\$.tw,kf.
 45 Education of Intellectually Disabled/
 46 education, special/
 47 speech therapy/
 48 language therapy/
 49 speech-language pathology/
 50 Sign language/
 51 Manual Communication/
 52 Nonverbal communication/
 53 Communication Aids for Disabled/
 54 Self-Help Devices/
 55 communication/
 56 (speech\$ or languag\$ or communicat\$ or sign\$ or nonverbal\$ or non-verbal\$ or cue\$).tw,kf. (5964622)
 57 or/41-56
 58 40 and 57
 59 exp Parents/ed
 60 Caregivers/ed
 61 ((parent\$ or maternal\$ or mother\$ or father\$ or paternal\$ or carer\$ or caregiver\$ or care-giver\$ or adult\$ or teacher\$ or therapist\$) adj3 (coach\$ or educat\$ or intervention\$ or learn\$ or program\$ or teach\$ or train\$)).tw,kf.
 62 ((parent\$ or maternal\$ or mother\$ or father\$ or paternal\$ or carer\$ or caregiver\$ or care-giver\$ or adult\$ or teacher\$ or therapist\$) adj3 (interact\$ or inter-act\$ or involv\$ or mediat\$ or respon\$)).tw,kf.
 63 or/59-62
 64 (naturalistic adj2 teaching).tw,kf.
 65 focus?ed stimulation.tw,kf.
 66 (milieu adj2 teaching).tw,kf.
 67 (responsiv\$ adj2 education).tw,kf.
 68 (responsiv\$ adj2 teaching).tw,kf.
 69 Hanen\$.tw,kf.
 70 or/64-69
 71 58 or 63 or 70
 72 randomized controlled trial.pt.
 73 controlled clinical trial.pt.
 74 randomi#ed.ab.
 75 placebo\$.ab.
 76 drug therapy.fs.
 77 randomly.ab.
 78 trial.ab.

79 groups.ab.
80 or/72-79
81 exp animals/ not humans.sh.
82 80 not 81
83 33 and 71 and 82

Appendix 2. Criteria for judging risk of bias

Random sequence generation (method of assigning participants to groups)

1. Low risk of bias: well-described, randomised process (e.g. coin toss; table of random numbers; computerised random number generator).
2. High risk of bias: non-random method (e.g. days of the week, alternate).
3. Unclear risk of bias: allocation is not described or description leads to uncertainty in quality of allocation, and possibility of bias.

Allocation concealment

1. Low risk of bias: we will rate concealment of allocation as adequate if random allocation schedules were developed by an independent researcher, and if allocation was recorded within opaque envelopes created by the independent researcher or the computerised allocation system was controlled by the independent researcher.
2. High risk of bias: providers of intervention undertake allocation or research team allocate participants and has access to participant characteristics.
3. Unclear risk of bias: methods of concealment are not described or description does not allow bias to be ruled out.

Blinding of participants and personnel

In the case of parent training interventions, neither the parent nor the trainer can be blinded to the type of treatment given. Blinding in studies in this review will refer to components of intervention being tested (e.g. video coaching; focus on an individual communication strategy). We will assess studies on an individual basis, however, it is likely that most studies will be at high risk of bias on this criterion due to the nature of the intervention.

1. Low risk of bias: parents and trainers are blinded to which of the two comparison treatments they have been allocated or are blinded to the exact feature of the intervention that is being tested.
2. High risk of bias: parents or trainers, or both, are not blinded to the exact focus of intervention being tested, or it is likely that blinding could have been broken during the study.
3. Unclear risk of bias: insufficient information provided to judge the knowledge of parents and trainers on the feature of the intervention being tested or the difference between individual treatments.

Blinding of outcome assessors

1. Low risk of bias: reports state that outcome assessors were blinded to group allocation.
2. High risk of bias: reports suggest that assessors are likely to know the group to which the participant was allocated (e.g. provided treatment, worked with person delivering treatment).
3. Unclear risk of bias: no information on blinding of assessors or role of assessors in allocation or treatment provision.

Completeness of data collection

We will report the numbers in each intervention group at each end point compared with total randomised participants and the reason(s) for attrition/exclusion if provided by study authors. If missing data are imputed we will consult a statistician about the appropriateness of the method used. If we retrieve and enter data into the review such re-inclusions in analyses will be reported.

1. Low risk of bias: no missing outcome data; or loss to follow-up is unlikely to be related to the true outcome or attrition is similar in both conditions and proportion of missing data (dichotomous outcomes) or effect size (continuous outcomes) are unlikely to have a clinically-relevant effect; imputation of missing data is judged to be appropriate.

2. High risk of bias: loss of participants to follow-up is likely to be related to the true outcome or is distributed unevenly across groups, or imputation of missing data is judged to be inappropriate. Studies showing uneven loss to follow-up will be considered separately in sensitivity analyses.
3. Unclear risk of bias: loss of participants to follow-up is not reported or insufficient information is provided to judge the reason for loss and judgement of low or high risk of bias.

Selective reporting

We will check published protocols to assess if all planned comparisons are reported. If protocols are not published we will contact study authors to ask if additional comparisons were planned.

1. Low risk of bias: all prespecified outcomes are reported.
2. High risk of bias: selective reporting of outcomes is evident.
3. Unclear risk of bias: not possible to judge if all planned comparisons have been reported.

Other sources of bias

We will describe any additional problems that may put a study at risk of bias.

1. Low risk of bias: study appears free from other sources of bias.
 2. High risk of bias: at least one important risk of bias (e.g. groups clearly different at baseline, children receiving direct therapy during the study period).
 3. Unclear risk of bias: insufficient information provided on which to judge other sources of bias.
- We will judge studies that score a low risk of bias for all criteria to be at low risk of bias overall. If studies are judged to be at low or unclear risk of bias for all criteria, we will judge them as being at unclear risk of bias overall. If studies are assessed to be at high risk of bias for one or more criteria, we will judge them to be at high risk of bias overall.

CONTRIBUTIONS OF AUTHORS

LP and JG conceived and designed the review.

LP wrote the protocol with drafts reviewed by KL and JG.

KL developed the search strategy with advice from LP and JG.

LP has overall responsibility for the review.

DECLARATIONS OF INTEREST

Lindsay Pennington (LP) is a speech and language therapist and senior lecturer at the Institute of Health and Society, Newcastle University, UK. She has led an early phase trial of parent-mediated intervention for children with cerebral palsy. She currently receives funding from Sparks, The Children's Medical Research Charity, to develop a smart phone app for use in parent-mediated therapy (14NCL01). This study does not fit the criteria for this review and will not be included. LP received payment of accommodation fees by Cerebral Palsy Alliance to present at a symposium on Early Intervention in 2016 at the International Cerebral Palsy Conference, Stockholm, Sweden.

Kate Laws (KL) is a highly specialist speech and language therapist, at the City Hospitals Sunderland NHS Foundation Trust, UK. She provides interventions for children with complex communication needs, including parent-mediated interventions. Her work on the development of this protocol was supported by a National Institute for Health Research Internship. KL's employer (City Hospitals Sunderland) receives a set fee from Health Education North, which is part of the NHS, to release her for 30 days as an internship with Dr Pennington, Health Education North Clinical Academic Training Programme.

Juliet Goldbart (JG) is Associate Dean for Research (Health, Psychology and Social Care Faculty) at Manchester Metropolitan University, UK. She currently receives funding from the National Institute for Health Research Health Services and Delivery Research (HS&DR) Programme, to conduct the following research: 'Identifying appropriate symbol communication aids for children who are non-speaking: enhancing clinical decision making'. The project addresses the tangentially-related research area of clinical decision-making

in augmentative and alternative communication. Her role is primarily in systematic reviews to inform the project. As such, she does not perceive any conflict of interest. JG's institution was paid by the International Society for Augmentative and Alternative Communication, Norway, for two lectures on early communication and evidence-based practice. JG receives royalties for Coupe-O'Kane and Goldbart (1997) 'Communication Before Speech'. JG declares the book focuses on children with profound disabilities rather than motor disorders and addresses assessment and generic strategies rather than specific interventions.

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